



First Identified Case in Literature: Association of Achalasia and Celiac Diseases

Akalazya ve Çölyak Hastalıkları Birlikteliği: Literatürde İlk Kez Tespit Edilen Olgu

Association of Achalasia and Celiac Diseases

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Özet

Çölyak hastalığında ösefagus sfinkterinde gevşeme ve mide boşalma zamanında uzama gibi gastrointestinal sistem problemleri görülmektedir. Akalazya hastalığı ösefagus peristaltizminde yokluk ve alt ösefagus sfinkterinde inkomplet gevşeme ile karakterize motor bir bozukluktur. Akalazya ve çölyak hastalıkları birlikteliği daha önce rapor edilmemiştir. Hastamızın son üç yıldır olan, büyüme ve gelişmesini etkileyen kusmaları mevcut idi. Hastamızda çölyak hastalığı tanısı serolojik ve histopatolojik olarak koyuldu aynı zamanda akalazya hastalığı ise ösefagoskopik muayene, üst gastrointestinal sistem kontrast çalışması ve ösefageal manometri ile koyuldu. Ösefageal balon dilatasyonu olguya uygulandı. Bu olgu ilginç birliktelik nedeniyle sunuldu.

Anahtar Kelimeler

Akalazya Hastalığı; Çölyak Hastalığı; Kusma

Abstract

Celiac disease has been shown to cause problems related to gastrointestinal system motility such as reduction of the esophageal sphincter pressure and prolongation of gastric emptying time. Achalasia disease is a motor disorder that is characterized by the absence of esophageal peristalsis and by incomplete relaxation of the lower esophageal sphincter. Association of achalasia and celiac diseases has not been reported yet. Our patient with growth and developmental retardation had vomiting effects which lasted for 3 years. Celiac disease was diagnosed serologically and histopathologically in our patient; we determined achalasia disease with esophagoscopy examination, upper gastrointestinal system contrast study, and esophageal manometry. Esophageal balloon dilatation was applied. This case is presented because of the interesting association between celiac disease and achalasia disease.

Keywords

Achalasia Disease; Celiac Disease; Vomiting

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Introduction

Celiac disease (CD) is an immune-mediated systemic disorder elicited by the ingestion of wheat gliadin and related prolamins, affecting individuals who are HLA DQ2 and/or DQ8 positive, and characterized by the presence of anti-tissue transglutaminase antibodies and/or gluten-dependent manifestations including enteropathy [1]. Achalasia disease (AD) is a motor disorder of the esophagus characterized by loss of esophageal peristalsis, increased low esophageal sphincter (LES) pressure, and absent or incomplete relaxation of the LES [2]. Celiac disease has been shown to cause problems related to gastrointestinal system motility such as reduction of the esophageal sphincter pressure and prolongation of gastric emptying time [3]. Association of achalasia and celiac diseases has not been reported yet. This case is presented because of this interesting association.

Case Report

A 12-year-old male patient was admitted to our clinic with complaints of vomiting. It was learned that the vomiting had been occurring for three years. The vomit contained solid and liquid food. On physical examination, the patient had a cachectic appearance. The other system examinations were normal. The patient had growth and developmental retardation [weight 28 kg (<3p) and height 138 cm (3p)]. Anti-tissue transglutaminase antibody (IgA and IgG) and anti-endomysium antibody were determined serologically (+). An esophagoscopy examination revealed lower sphincter constriction without stenosis. Esophageal mucosa was normal. His endoscopic and histopathological examination of the duodenum was compatible with celiac disease (Marsh 3c). On the upper gastrointestinal system contrast study of the case, the esophagus was severely dilated with distal bird's beaking. Passage into the stomach was also slow and limited (Figure 1). Esophageal manometer of the case was

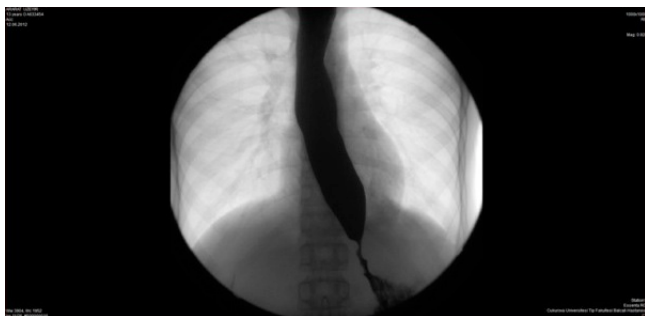


Figure 1. Esophagus was severely dilated with distal bird's beaking.

compatible with AD and esophageal balloon dilation was applied. The case benefited from treatment, but vomiting began again at the 10th day. Balloon dilation was applied again and was followed up in the 2nd month. His swallow function is better than before and he is not vomiting.

Discussion

Celiac disease is a disorder that is characterized by a de-regulated immune response to ingested wheat gluten and related cereal proteins in susceptible individuals. Clinical features of celiac disease vary considerably. Intestinal symptoms are common in children; failure to thrive, chronic diarrhea, vomiting, abdominal distention, muscle wasting, and anorexia are present in most cases. AD is a motor disorder that is

characterized by the absence of esophageal peristalsis and by incomplete relaxation of the lower esophageal sphincter [4]. The most common symptoms in children and adolescents are vomiting, progressive dysphagia, and weight loss [5].

Our patient with growth and developmental retardation had vomiting which lasted for 3 years. Celiac was diagnosed serologically and histopathologically in our patient; we determined AD with esophagoscopy examination, an upper gastrointestinal system contrast study, and esophageal manometer.

Triple A syndrome, Rozycki syndrome, Chagas' disease, Sarcoidosis, Down syndrome, Paraneoplastic syndromes, Hodgkin's disease, Cerebellar ataxia, and Myoneural disorders have been shown to be associated with AD [2]. But there is no association between celiac disease and AD in the literature.

Celiac disease is an autoimmune disorder. Some diseases, including many with an autoimmune pathogenesis, are found with a higher than normal frequency in celiac patients [1]. An autoimmune etiology for AD has been considered because of the presence of neural inflammation in the absence of conclusive evidence of infection. Studies have demonstrated inflammatory cell infiltrate of the myenteric plexus in 90%-100% of esophageal specimens from AD patients [6]. Storch et al. suggested a role of autoimmunity by demonstrating a higher prevalence of anti-myenteric autoantibody in AD (64%) [7]. Verne et al. tried to demonstrate the presence of regional and cellular specific antibody in AD [8]. Although genetic predisposition and autoimmunity are important in both diseases, there is no clear data to explain the cause of these diseases seen together. In addition, the fact that this association is rare in the literature suggests that it might be a coincidence.

In patients with celiac disease, increase in gastrointestinal motility disorder, and frequent reflux have been demonstrated. In the cases with celiac, however, no presence of AD is observed in the literature. This case is presented because of this interesting association.

Competing Interest

The authors declare that they have no competing interest.

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